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The Slav-cleft: A three-center study of the outcome of treatment of cleft lip and palate. Part 1: Craniofacial morphology

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ABSTRACT

Results of a comparison of the outcomes of treatment of cleft lip and palate can be affected by growth characteristics of populations from which subjects with the clefts are derived. Moreover, conventional cephalometric techniques used in cleft studies for analysis of facial morphology provide only a partial description of shape and are confounded by biases regarding the reference structures. In this retrospective comparison, craniofacial morphology of preadolescent patients with unilateral cleft lip and palate treated in Warsaw (n = 35, age = 10.6 years, SD = 1.2), Prague (n = 38, age = 11.6 years, SD = 1.4), and Bratislava (n = 26, age = 10.5 years, SD = 1.6) were evaluated on cephalograms with the cephalometric method used in the Eurocleft study and geometric morphometrics. We found that patients treated in Warsaw showed slightly more favorable outcomes than in Prague and Bratislava. The differences were related primarily to the position of maxillary alveolar process, cranial base, mandibular angle, and soft tissues. Although no association between a component of treatment protocol and the outcome was found, it is possible that organizational factors such as participation of high-volume, experienced surgeons contributed to these results.

1. Introduction

An evaluation of craniofacial morphology in patients with orofacial clefts is part of a comprehensive assessment of treatment outcomes. Usually, it is carried out comparing the effectiveness of different methods of treatment. However, problems can arise when the comparison is performed in an international setting and patients treated in different cleft centers have also different ethnic backgrounds. In such situations, morphological differences between background populations can affect the findings. This issue was discussed in relation to the Eurocleft study, a large intercenter comparison of treatment outcomes for cleft lip and palate in northern and western Europe (Trenouth et al., 1999). Trenouth et al. (1999) compared facial growth of 9- and 12-year-olds without any cleft from several countries including Norway and England. They found that for both ages, the maxillary convexity described by the sn-ss (SNA) angle, increased by 1.7° for young persons from Norway, whereas it decreased by 2.6° for boys from Manchester, England. Thus, the maxilla was considerably more prominent in 12-year-old Norwegians than for their English peers. This finding, in turn, could affect the outcome of comparing Norwegian and English patients with clefts, because any difference in maxillary prominence detected in children with the cleft could be partly a result of distinctive craniofacial growth in background populations. A similar challenge was encountered while interpreting the results of the comparison between cleft centers in Warsaw and Oslo (Fudalej et al., 2015); a more prominent maxilla found in Norwegians could have resulted from differences in facial growth trajectories between

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the two populations or from more favorable treatment in the case of the Oslo group. A comparison of samples with the same ethnic background can overcome this problem to a certain degree.

The neighboring countries of Poland, the Czech Republic, and Slovakia, are predominantly Slavic. The term *Slavic* describes the largest Indo-European ethno-linguistic group in Europe that shares a long-term cultural continuity and speaks a set of related languages. Present-day Slavs are classified as West, East, or South Slavs, with Poles, Czechs, and Slovaks belonging to the West Slavic group (http://www.britannica.com/topic/Slav). Although the common ethno-linguistic origin of Poles, Czechs, and Slovaks does not guarantee that the craniofacial morphology of these populations is identical, the anthropometric research has revealed some differences between Slavs and other ethnic groups (e.g., Anglo-Saxons, Latinos, etc.) (Kolar, 1987). For example, despite a large within-group variability of the cephalic index, it has been demonstrated that Anglo-Saxons are significantly more scaphocephalic than Slavs. Other studies also imply that the craniofacial morphology of Slavs is different than for those from other ethno-linguistic groups. Ross (2004) evaluated craniofacial variation in Croatians, Bosnians, Macedonians, Greeks, and white Americans living in the 20th century. She found a marked differentiation among Balkan groups, which was ordered relative to ethno-linguistic ancestry – The Mahalanobis distance (D^2) between mean craniofacial shapes of Bosnians and Croats (both groups are relatively homogenous and historically to originate from the same Slavic ancestry) was 4.5, whereas D^2 between Bosnians and white Americans was 11.1, and between Bosnians and Greeks was 19.2. This indicates that ethno-linguistic *distance* is associated with the degree of differences in craniofacial shapes. Furthermore, genetic differences between European populations can be related to ethno-linguistic background. For example, Barbujani and Sokal (1990) found that out of 33 gen-frequency boundaries in Europe, 31 were coincident with linguistic boundaries. Although no direct evidence is available, it seems sensible to assume that craniofacial morphology of Poles, Czechs, and Slovaks is quite comparable.

Of the three cleft centers participating in this study, the Prague center did not compare the effects of its treatment protocol with the outcomes achieved in other centers, whereas the Bratislava center participated in one comparative investigation (Koželj et al., 2012). In contrast, the cleft team from Warsaw Institute of Mother and Child had participated in several international comparisons (Fudalej et al., 2009a, 2009b, 2015) and its outcome was found to be relatively advantageous. Assuming similarity of facial form in Polish, Czech, and Slovak populations, it seems appropriate to evaluate morphology of the craniofacial region in patients with orofacial clefts treated in Warsaw, Prague, and Bratislava. In such a comparison, the Warsaw group would serve as a reference sample. Therefore, the objective of this study was to compare facial morphology in a sample of patients with complete unilateral cleft lip and palate treated in three centers (Warsaw, Prague, and Bratislava) using different surgical protocols. The H_0 hypothesis is that facial morphology in all groups is comparable.

2. Materials and methods

2.1. Subjects

Preadolescent children with cleft lip and palate (CLP) from three Central European cleft centers – Warsaw (Poland), Prague (Czech Republic), and Bratislava (Slovakia) – were selected for this retrospective study of facial morphology. The inclusion criteria were complete unilateral cleft lip and palate (CUCLP) operated on at the respective center, and lateral cephalograms taken at about 10 years of age. The exclusion criterion was CUCLP associated with other syndromes.

The Warsaw (W) group comprised 35 subjects (25 boys and 10 girls) born between July 1993 and January 1996. They were consecutively operated on by a single experienced surgeon at the Warsaw Institute of Mother and Child between May 1994 and August 1996. In all subjects, the CUCLP was corrected with a one-stage surgical protocol. The details of the protocol were described by Fudalej et al. (2009a). Radiographic assessment was carried out at a mean age of 10.6 years (SD = 1.2, range = 8–13.6).

The Prague (P) group comprised 38 subjects (27 boys and 11 girls) taken from a series of 77 patients born between the years 2000 and 2003. They were treated consecutively by the cleft team at the Center for Treatment of Craniofacial Anomalies in Prague. The CUCLP was closed in 2 stages; closure of the lip was done at 7.3 months (SD = 5.5, range = 3.9–35.2) using the Tennison–Randall technique, whereas closure of the hard and soft palate was performed at 35.5 months (SD = 6.4, range = 18.4–54) using the Wardill–Kilner method (in some patients, the Wardill–Kilner method was combined with vomerplasty).

Five surgeons were involved in the closure of the CUCLP. No infant orthopedics (IO) was carried out.

Radiographic assessment was carried out at mean age of 11.6 years (SD = 1.4, range = 8.8 to 14.4).

The Bratislava group (B) comprised 26 subjects (19 boys and 7 girls) taken from a series of 44 patients born between the years 2000 and 2005. They were consecutively treated by the cleft team at the Clinic of Plastic and Reconstructive Surgery, Comenius University in Bratislava. The CUCLP was closed in 2 stages; closure of the lip was done using the Millard technique at 4.6 months (SD = 1.8, range = 2.4–8.5), whereas closure of the palate was performed using the Wardill–Kilner method (in some patients, the Wardill–Kilner method was combined with vomerplasty) at 12.4 months (SD = 6.4, range = 7.6–42.7). Five surgeons were involved in the treatment; a single surgeon operated on 18 patients, and 4 surgeons operated on the remaining 8 patients. Infant orthopedic treatment was performed on 23 patients (3 patients did not receive IO). Radiographic assessment was carried out at mean age of 10.5 years (SD = 1.6, range = 7.6–13.8).

A summary of the Warsaw, Prague, and Bratislava protocols is provided in Table 1.

2.2. Methods

Craniofacial morphology was analyzed on lateral cephalograms taken in centric occlusion using two methods: (1) the cephalometric protocol applied previously in the Eurocleft study (Brattström et al., 2005), and (2) geometric morphometrics (GM). In both methods, scans of cephalograms (or digital cephalograms) were downloaded into the Viewbox software, version 4 (dHAL software, Kifissia, Greece), and 27 landmarks (15 for hard tissues and 12 for soft tissues; Fig. 1) were identified by one investigator (P.F.). In line with the Eurocleft cephalometric protocol, 13 angular and 2 ratio variables were calculated to compare groups. In contrast to the cephalometric protocol, geometric morphometrics used generalized partial least-square Procrustes superimposition of the same sets of landmarks to extract coordinates of craniofacial shape, which were subsequently analyzed (Halazonetis, 2004).

2.3. Statistical analysis and method error

Descriptive statistics (means and standard deviation) were computed for each group. One-way analysis of variance (ANOVA) with Tukey–Kramer post-hoc pairwise tests was carried out to identify intergroup differences for angular and ratio variables. To

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